

## Case Report

### A Case Report on Unilateral Double Testicles

AC Paul<sup>1</sup>, MK Hassan<sup>2</sup>, MJ Alam<sup>3</sup>, SK Biswas<sup>4</sup>, AK Saha<sup>5</sup>, NI Mridha<sup>6</sup>.

#### Abstract

A unilateral double testicle is a rare anomaly characterized by migration of one testicle towards the opposite inguinal canal. The commonest erratic development is the more or less incomplete descent of the testicle along the normal route of descent, which is known as cryptorchidism. In ectopia of the testicles, as opposed to cryptorchidism, the displaced testicle does not descend along the usual route but as it migrates downwards it moves into an entirely abnormal position. Usually the migrating testicle remains on its own side of the body but may end up in an unusual position e.g. in the superficial tissue of the inguinal region above the external ring, in the area of the base of the penis, in the upper part of the thigh, in the region of perineum or in the pelvic cavity. In this case, the right and left testicles were found to descend together on the right side, whereas the left side of the scrotum was entirely empty. The case was treated with 'Bilateral Transeptal Subdartos Orchidopexy'. The case reported here is evidently one of extreme rarity as there are about 148 reported cases since the first described by Von Lenhossek in 1886.

**Key words:** Cryptorchidism, Testicle, Ectopia, Orchidopexy, CTE.

#### Case report:

A one year old boy presented with a painful swelling in the right groin and vomiting for 6 hours with a history of empty scrotum on the left side since birth. Physical examination showed a right sided obstructed inguinal hernia together with a palpable retractile right testicle and a non palpable left testicle and spermatic cord. Suspecting a right sided obstructed inguinal hernia and left sided cryptorchidism, he was submitted to emergency operative treatment under general anesthesia after adequate resuscitation.

Operative findings as related to this unusual condition are described now. On inguinoscopy, a redundant tunica vaginalis was discovered which extended for some distance proximally along the cord. During separation

1. Dr. Amal Chandra Paul, MS (Paediatric Surgery), Assistant Professor, Dept. of Paediatric Surgery, FMC, Faridpur.
2. Dr. Md. Kamrul Hassan, MS (Paediatric Surgery), Assistant Professor, Dept. of Paediatric Surgery, FMC, Faridpur.
3. Dr. Md. Jahangir Alam, DA, Assistant Professor, Dept. of Anesthesia, FMC, Faridpur.
4. Dr. Swapan Kumar Biswas, FCPS (Surgery), MRCS (Edin), Assistant Professor, Dept. of Surgery, FMC, Faridpur.
5. Dr. Alope Kumar Saha, FCPS (Paediatrics), Assistant Professor, Dept. of Paediatric, FMC, Faridpur.
6. Dr. Nazrul Islam Mridha, MBBS, Registrar, Dept. of Paediatric surgery, FMC, Faridpur.

#### Address of correspondence:

Dr. Amal Chandra Paul, MS (Paediatric Surgery), Assistant Professor, Dept. of Paediatric Surgery, FMC, Faridpur. Phone: +088-01819489871, E-mail: msamal43@yahoo.com

of the tunica vaginalis, it was found that the boy had two testicles on the same side of equal appearance, separated epididymis and vasa deferentia. The mesorchia was fused with the peritoneum. Both testicles were delivered into the inguinal wound (Fig.1). Both spermatic cords were followed to the right deep inguinal ring where they entered the abdomen. Isolation of the hernial sac was carefully done in a regular way without any injury to the spermatic cords and spermatic vessels.



Fig.1: Both testicles at inguinal incision



Fig.2: Follow up on 28th POD.

Although each testicle had a separate spermatic cord, there was a common tunica vaginalis enclosing the testicles. At the time of operation the left scrotum was examined and no evidence of testicle could be found there. The right external ring area was widened in order to deliver both testicles along with the spermatic cords to the right scrotal sac. Alignment of the cords and blood vessels was maintained. The right one was kept in the right scrotum and a transeptal subdartos tunnel was made under the median scrotal raphe. The other testicle was pulled to the left subdartos area. Both testicles were observed for ten minutes for any vascular impairment. Good maintenance of vascularity was observed. Then right and left orchidopexy was performed. Post operative follow up showed good positioning of both testicles and both scrotal sacs appeared to be normal, separated by a clear median raphe (Fig. 2).

### Discussion:

There are about 148 cases of unilateral double testicles described since 1886, when Von Lenhossek reported a necropsy finding<sup>1</sup>. The largest incidence is found in Europe and Japan. Sixty cases having been published in Japanese literature since the first report in 1912, by Iwasaki<sup>2</sup>. Unilateral double testicles are also termed as Crossed Testicular Ectopia (CTE). Demographic statistics of the United Nations, the estimation of global incidence of CTE is about 1:4 million<sup>3</sup>. There is no statistical difference in regard to the affected side, and over the last 20 years the mean age at diagnosis has been 9.3 years. One case of family incidence was reported by Stauber, in two brothers with CTE and persistent mullerian remnants<sup>4</sup>.

Our child presented a clinical picture analogous to the most frequently found in CTE. This patient presents with obstructed inguinal hernia with ipsilateral palpable testis and an impalpable testis on the other side. As surgery is indicated for the obstructed hernia, and due to the rarity of unilateral double testicles diagnosis is made at operation.

There are some differences among the various cases of CTE, which have produced several theories to explain the genesis of this rare entity<sup>5-9</sup>. Many authors propose that abnormal or absent gubernaculum could be important factors, although it has been demonstrated normal testes migration after gubernaculum ablation in animal fetuses<sup>10</sup>. Most authors agree that each testis is formed on different sides, and somehow one crosses toward the opposite side in the major part of the migration trajectory. Many believe mechanical causes like internal inguinal obstruction, absent peritoneum vaginalis process, absent gubernaculum, mesorchia adhesions, and duct or gonads fusion, are certainly relevant factors.

Thevathasan postulated a classification of CTE considering the eventual etiology<sup>11</sup>. There is a simple

classification into three types, (Type-I: Simple CTE, associated to inguinal hernia alone, Type-II: CTE associated to persistent mullerian remnants, Type-III: CTE associated to other anomalies) based upon the objective presence of associated anomalies, which would imply distinct therapeutic approaches.

In the evidence of unilateral double testicles at the operation, the approach depends on the operatory findings, Type I should be treated with dissection and high ligation of the hernia sac, dissection and isolation of both cords and vas deferens and orchidopexy<sup>12</sup>. If spermatic cord length is good, as in our case, it is recommended to fix the ectopic testis in the opposed scrotal pouch by transeptal technique<sup>13</sup>. If the spermatic cord is short, one can perform testis transposition to the other groin through contralateral inguinoscrotal approach, with or without laparotomy, by intra or extra peritoneal approach, or by staged orchidopexy. Type-II: besides the steps mentioned above, in the presence of mullerian remnants, their ablation is not obligatory, and just a segmentary resection may be done to provide pathological study. Type III: In the evidence of other anomalies, treatment should be appropriate to each case. Genetic evaluation shall be performed methodically. Despite association of 1-2% of high urinary tract anomalies, it is recommended urographic investigation in all cases of crossed testicular ectopia.

### Conclusion:

A one year old boy presented with a clinical condition of an obstructed inguinal hernia on the right side and contralateral impalpable testicle. The boy was operated as an emergency case on the basis of clinical diagnosis and diagnosis of unilateral double testicles was made at operation. In this case both testicles appeared to be normal in contrast to other reported cases<sup>14</sup>. Postoperative follow up revealed a normal well developed scrotum with a defined median raphe. An experienced sonologist may detect the finding preoperatively. Treatment includes transeptal orchidopexy, transeptal orchidopexy with some modification (in this case), extraperitoneal transposition of the testicles as well as search for mullerian remnants and other anomalies. Long term follow up is necessary because of the risk of becoming malignant.

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